Chronic Melioidosis in a Patient with Cystic Fibrosis

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Burkholderia pseudomallei, the causative agent of melioidosis, is endemic in Southeast Asia and northern Australia, where it can be found in soil and surface water. We report a case of chronic pulmonary melioidosis in a patient with cystic fibrosis who had traveled to an area where B. pseudomallei is endemic.

CASE REPORT

The patient was a 38-year-old white male with cystic fibrosis (CF) who had undergone middle-lobe resection of the lung for bronchiectasis at the age of 17. During the following 10 years he has had only mild pulmonary symptoms. Since his lung function worsened during each winter season he started to spend the winter months in warmer areas: 1989 and 1990 in Indonesia and 1990 and 1991 in Mexico, the Dominican Republic, California, and Costa Rica. On returning from Thailand in the spring of 1992 he was severely ill with fever, cough, and malaise and was admitted to Freiburg University Hospital. The patient's sputum sample yielded a gram-negative rod that was identified by API 20NE (code 1140474) as a Pseudomonas sp. Since then he had 13 documented exacerbations with gram-negative rods that were reported to be either Pseudomonas sp., Pseudomonas aeruginosa, or Burkholderia cepacia. In each case, he was treated for a maximum of 2 weeks with anti-pseudomonas antibiotics (ceftazidime, piperacillin-tazobactam, or piperacillin with or without aminoglycosides or ciprofloxacin) administered intravenously (i.v.) with only moderate effect. The isolated strains were susceptible to the drugs used with the exception of aminoglycosides. A detailed travel history was not documented, and B. pseudomallei and melioidosis were never suspected by the microbiologists and clinicians, respectively, associated with this case.

In November 1998 a new screening plate for Burkholderia species (3) had been implemented in the diagnostic laboratory, and a sputum plate was sent to a reference laboratory (Munich, Germany). There, as in our own laboratory, Burkholderia pseudomallei was identified on the basis of biochemical tests (API 20NE code @48h 1156577) and by means of 16S rRNA gene sequencing and comparison to the type strain 1026b (GenBank accession number U91839). Moreover, the isolate gave a positive reaction in a recently developed B. pseudomallei-specific latex agglutination test (11) which was not available in the diagnostic laboratory at the time of isolation. The isolate was susceptible to ceftazidime, imipenem, trimethoprim-sulfamethoxazole, tetracycline, and ampicillin-sulbactam and was resistant to all aminoglycosides, fosfomycin, and colistin. Bacte-

Melioidosis, a tropical disease caused by B. pseudomallei, may present itself in a variety of unusual ways, such as neck lumps (5), brain abscess (6), or infection of the placenta, resulting in transplacental mother-to-child transmission (J. M. Orendi, F. C. H. Abbink, and A. J. de Beaufort, abstr. MoP 130, Clin. Microbiol. Infect. 6(Suppl. 1):43, 2000). The potential for diagnostic confusion of B. pseudomallei infection with other diseases may be high in such cases. We report here the first case of chronic B. pseudomallei infection in a patient with CF. This case also highlights the need for a careful microbiological diagnosis to discriminate the expected from the unexpected.

In contrast to antipseudomonas therapy in CF, the optimal combination therapy for severe melioidosis has not been conclusively determined (10). It remains unclear whether an earlier recognition of the causative agent in this patient, resulting in an initial i.v. therapy with ceftazidime or carbopenems (9), followed by long-term maintenance with amoxicillin-clavulanate, would have had a positive effect on the course of the disease. It has been reported that B. pseudomallei can be identified by a combination of the commercial API 20NE biochemical kit and a simple screening system involving Gram stain, the oxidase reaction, resistance to colistin and gentamicin, and typical growth characteristics on Ashdown medium (2). However, the need to perform repeated testing of some strains with
the API 20NE kit in order to get the correct identification has
been reported (2), as well as misidentifications of B. pseudoma-
llei as other species (4). A review of bacteriological reports in
our case revealed that the API 20NE profiles of some former
isolates were recorded after 24 h. Dance et al. have reported
that 48 h of incubation of the panel is crucial for a correct
identification of B. pseudomallei (2). We think it is reasonable
to confirm a presumptive identification of B. pseudomallei
based on biochemical results and resistance pattern by using
serological methods such as the monoclonal antixoplyasac-
charide antibody-based B. pseudomallei-specific latex aggluti-
nation test (11). This might be especially true for laboratories
where only occasional imported strains have to be identified. This
case also demonstrates that testing patient sera for B. pseudo-
mallei-specific antibodies might be a helpful tool for confirm-
ing the diagnosis.

Recently, imported melioidosis has been reported in pa-
tients with various underlying diseases (1), including one CF
patient, but no history or clinical details were provided. It is
highly suggestive that our patient became infected in Thailand,
although we cannot completely rule out the possibility of a
primary infection occurring during his visits to countries where
B. pseudomallei is also endemic before he went to Thailand.
Colonization and infection with Burkholderia species belonging
to the B. cepacia complex and with B. gladioli is a well-recog-
nized problem in CF patients and is associated with decreasing
lung function and disease progression (7). One might speculate
as to whether the changes in CF lungs predispose individuals
to infection with B. pseudomallei. As a result of better manage-
ment options for CF patients, the quality of life and life ex-
pectancy is increasing, and therefore patients’ travel activities
may be increasing as well. Detailed history taking is crucial,
and one should not be biased by the underlying disease when
interpreting the biochemical results and susceptibility data.
Awareness of melioidosis should be heightened since, as this
case demonstrates, diagnosis may be delayed, therapy is diffi-
cult, and the outcome uncertain.

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